

Cotard's syndrome: Two case reports and a brief review of literature

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ABSTRACT

Cotard's syndrome is a rare neuropsychiatric condition in which the patient denies existence of one's own body to the extent of delusions of immortality. One of the consequences of Cotard's syndrome is self-starvation because of negation of existence of self. Although Cotard's syndrome has been reported to be associated with various organic conditions and other forms of psychopathology, it is less often reported to be seen in patients with catatonia. In this report we present two cases of Cotard's syndrome, both of whom had associated self-starvation and nutritional deficiencies and one of whom had associated catatonia.

Key words: Catatonia, Cotard's syndrome, depression

Introduction

Cotard's syndrome is a rare neuropsychiatric condition characterized by anxious melancholia, delusions of non-existence concerning one's own body to the extent of delusions of immortality.^[1] It has been most commonly seen in patients with severe depression. However, now it is thought to be less common possibly due to early institution of treatment in patients with severe depression with psychotic symptoms.^[2] Cotard's syndrome is very infrequently reported in the setting of catatonia.^[3-6] There are occasional reports of Cotard's syndrome being accompanied by nutritional deficiencies. In this report we present two cases of Cotard's syndrome, both of whom had associated self-starvation and nutritional deficiencies and one of whom had associated catatonia and present a brief review on the topic.

Case Report

Case 1

Mr. B, 65-year-old retired teacher who was pre-morbidly well adjusted with no family history of mental illness, with personal history of smoking cigarettes in dependent pattern for last 30 years presented with an insidious onset mental illness of one and half years duration precipitated by psychosocial stressors. His symptoms were initially characterized by sadness of mood with early morning worsening, poor socialization, anhedonia, marked anxiety, decreased sleep and appetite, ideas of worthlessness, hopelessness, sin and guilt. As his symptoms progressed further he developed delusions of catastrophe, nihilism, poverty and persecution. Nihilistic delusion involved a description of everything coming to an end. He would verbalize that his organs are no more working, his brain has stopped functioning, and his house has developed cracks and is going to fall down. About a month prior to being admitted to our inpatient unit, he attempted suicide by hanging himself, but was saved. His suicide note revealed that he wanted to kill himself as he feared spreading a deadly infection to the villagers who resultantly might suffer from cancer. Later he started believing that he was dead and would not eat anything, due to this lost significant amount of weight. Over the next 2 months he attempted to end his life two

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more times. Following one of such suicide attempt he was brought to our outpatient unit and was admitted. Medical history and physical examination revealed presence of hypertension, malnutrition, nutritional large fiber neuropathy, benign prostatic hypertrophy, chronic obstructive pulmonary disease and chronic otitis media. Mental state examination revealed sad affect, marked agitation, ideas of hopelessness, delusion of catastrophe, delusion of guilt, sin and nihilism (of being dead). He lacked insight into his illness. On the basis of the history and mental status examination a diagnosis of severe depression with psychotic symptoms was made. His Hamilton Depression Rating Scale (HDRS) score at time of admission was 34. His investigations including thyroid function test and MRI brain did not reveal any abnormality. His nutritional deficiencies were addressed. His depression was successfully treated with bilateral modified electro-convulsive treatment (ECT) using thiopentone for induction and succinylcholine for muscle relaxation. The patient was administered ECT three times (Monday, Wednesday and Saturday) weekly and in total received 9 ECT. Additionally he was treated with escitalopram 15 mg/day and olanzapine 10 mg/day which were started few days prior to starting ECT and were continued during the period when patient was receiving ECT. His symptoms resolved completely over the period of 7 weeks and his HDRS at the time of discharge was 1. He maintained improvement after discharge from the inpatient unit.

Case-2

Ms A, 62-year-old homemaker has been suffering from bipolar affective disorder for 35 years. She presented to us with a relapse which started about 7-8 months ago characterized by symptoms of depressed mood, anxiety, decreased socialization, loss of appetite and subsequent weight loss, decreased interest in household, poor self-care, ideas of worthlessness, pessimistic views of future, forgetfulness and psychomotor retardation. Later on she developed nihilistic delusions in which she negated the existence of her body parts and existence of her family members. Negation of existence of family members was not accompanied by any persecutory delusion or misidentification delusions. She also believed that she has been rendered very poor. She would say that their house may collapse any day and the occupants will die. Over the period she started refusing food as she considered herself to be dead. Her illness further progressed and about 4 weeks prior to admission to inpatient unit she developed mutism, refusal to eat, rigidity in all the limbs and fecal and urinary incontinence. She was brought to the emergency room of our institute with aforementioned symptoms. Her physical examination revealed mild pallor, dehydration,

bilateral pitting edema and she was emaciated. On mental state examination she had sad affect, mutism, rigidity in all the four limbs and negativism. Her investigation showed hemoglobin of 10 gm/dl, hypoproteinemia and hypoalbuminemia. Further investigation revealed low vitamin D₃ levels and the magnetic resonance imaging of brain showed cerebral atrophy with mild subdural effusion. On the basis of history and examination the diagnosis of bipolar affective disorder current episode severe depression with psychotic symptoms, malnutrition and anemia were considered. She was given a trial of intravenous lorazepam but to no relief. Therefore, decision to start ECT was taken. She was given bilateral modified ECT using thiopentone for induction and succinylcholine for muscle relaxation. Her catatonic and depressive symptoms melted away with a course of 8 ECTs and her score on Bush Francis Catatonia Rating Scale score reduced from 14 to 0 at end of 8 ECTs. Her nutritional deficiencies were addressed appropriately. Later on she was started on mood stabilizers, olanzapine and fluoxetine.

Discussion

In one of his first description of psychopathology in 1880 Cotard described a case of "*delire hypochondriaque*" in a middle-aged female who negated the existence of her brain, nerves, chest or entrails and considered herself to be made of skin and bones. She also negated the existence of god and devil. Few years later, Cotard, introduced the term "*Delire des negation*" for patients who have a negating attitude of denying the existence of self or world conforming to the characteristics of delusions.^[7] Cotard reported it to be a variety of delusion which was commonly associated with severe depression, marked psychomotor retardation, presence of anxiety symptoms and other depressive symptoms.^[8,9] Cotard considered it to a type of depression and categorized it as *lypémanie*, a kind of psychotic depression described by Esquirol.^[10]

The term Cotard's syndrome was coined by Emil Regis and was later promoted by several others to describe patients presenting with anxious melancholia, delusions of absence of organs, negation, damnation and immortality.^[10]

In a review of literature, Berrios and Luque^[7] did an analysis of 100 cases of Cotard's syndrome and found depression to be most common diagnosis seen in patients presenting with Cotard's syndrome. In terms of phenomenology, nihilistic delusions concerning body and existence to be most frequent. Other descriptions

include negation of mind, brain, intellect, denial of pregnancy, delusional paralysis and cosmos or the world. The nihilistic delusion may also include denial of existence of various aspects of body or self, i.e. personal space like denial of being alive, non-existence of one's soul, personal name/age, self-movement/capacity to walk, capacity to eat, existence of function of heart/brain/liver/intestines/limbs etc., The aspects of extra-personal space denied in nihilistic delusions may include existence of world, marriages, parents or children.^[10] The first case of ours negated the functioning of his brain to start with and later negated his own existence. In the second case also the patient negated her own existence and it was associated with negation of existence of other family members.

Cotard's syndrome is in general reported to be more common in females and older age groups with rare occurrence in adolescents.^[11] Both of our patients were older than 60 years of age.

In terms of medical conditions Cotard's syndrome has been shown to be associated with syphilis, typhoid fever,^[12] migraine,^[13] epilepsy,^[14] cerebral trauma,^[8] cerebral arterio-venous malformation and epilepsy,^[14] arterio-venous malformation and multiple sclerosis,^[15] cerebral infarction,^[14] superior sagittal sinus thrombosis,^[16] brain tumors,^[17] temporal lobe epilepsy,^[14,18] limbic epileptic insults,^[18] Laurence-Moon/Bardet-Biedl syndrome,^[19] Parkinson's disease,^[20,21] brain injury,^[8,22-24] arachnoid cyst,^[25] noninfectious complication of heart transplantation,^[26] as a consequence of an adverse drug reaction to acyclovir and its prodrug valaciclovir,^[27] and in herpetic^[28] and non-herpetic encephalitis.^[29]

In term of psychiatric conditions Cotard's syndrome has also been reported in patients with severe mental retardation^[30] and postictal depression.^[31]

In terms of association with other phenomenological descriptions, case reports have also described the occurrence of Cotard's syndrome with hydrophobia,^[32] Lycanthropy,^[33] folie a deux,^[34] Capgras delusion,^[34-39] Capgras and Fregoli delusion,^[40] Odysseus syndrome,^[41] depersonalization disorder,^[29] catatonia,^[3-6] voluntary starvation^[42] and Koro like syndrome.^[43] In both our cases, voluntary starvation secondary to existence of self was noted.

There are only four reports of Cotard's syndrome being accompanied by catatonia.^[3-6] As with other reported cases, catatonia in our patient also came much later than the nihilistic delusions. Among the four cases described in the literature, only one

case was noted in a patient with bipolar disorder, as observed in our case.

On the basis of exploratory factor analysis of data of 100 cases, Berrios and Luque^[10] have reported three types of Cotard' syndrome: Psychotic depression (patients with melancholia and nihilistic delusions), Cotard type 1 included the pure forms of nihilistic delusions without affective symptoms and Cotard type 2 with a mixed group of symptoms of anxiety, depression and auditory hallucinations.^[10] Description of both of our cases fits into the psychotic depression subtype.

It is generally said that the symptoms of Cotard's syndrome develop over the period. Taking this into account three stages of development of Cotard's syndrome, namely germination stage, blooming stage and chronic stage have been proposed.^[44] The initial germination or prodromal stage is characterized by hypochondriasis and cenesthopathy. The blooming stage involves development of full symptoms of nihilistic delusions and chronic stage involves chronic changes in mood and systematization of delusions. In both our cases, symptoms evolved slowly and can be said to have followed these stages of development of psychopathology.

In terms of neurobiological evidence,^[1,24] many studies suggest lack of gross abnormalities in the neuroimaging. However, studies which have found neurobiological changes suggest involvement of fronto-temporo-parietal circuitry in the pathophysiology of Cotard's syndrome.^[1] In terms of personality attributes, studies suggest that patients with a more internal attributional style are more prone to develop Cotard's syndrome.^[24,28,29] In terms of neurophysiological changes some authors suggest lack of differential autonomic response to anything and reduction of affective response to stimuli in patients with Cotard's syndrome.^[1] Role of cultural factors in shaping the psychopathology in patients with Cotard's syndrome has also been proposed.^[45]

In terms of treatment of Cotard's syndrome,^[1,25] ECT has been reported to be useful although there are case reports of beneficial effect of antidepressant alone, antipsychotic alone or a combination of both.^[1] In both of our cases, ECT was found to be beneficial.

Both of the cases mentioned here, in due course of their illness, started believing that their body parts did not exist and thus reduced food intake which was evident as nutritional deficiencies. This can be partly due to severe depressive illness they suffered from and can also be attributed to the psychopathology of denial of

their existence. The second patient reported by us went on to develop catatonia in due process of progression of illness.

Our cases highlight the existence of phenomenology of Cotard's syndrome in patients with severe depression, especially those who present to treatment late. In the presence of Cotard's syndrome ECT is a useful treatment option.

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